CASE REPORT

Uterus Didelphys and Double Vagina with Delivery of a Normal Infant from Each Uterus

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THE incidence of congenital abnormalities of the uterus associated with pregnancy varies between 0.118%[1] and 0.3%[2] of deliveries.

An individual physician's experience with these abnormalities is limited. Kamm and Beernik[3] stated that because of this it is worth while for many physicians to report individual cases so that a collective review in the future would be meaningful.

In the following communication the case described is that of a woman with a double uterus and vagina who, after several miscarriages, was delivered of a normal infant by cesarean section from one uterus and subsequently of a normal infant from the other uterus, again by cesarean section.

Mrs. C.V.S., aged 19, was admitted to Highland View Hospital, Amherst, on June 11, 1962, complaining of lower abdominal cramps and bright vaginal bleeding. A tentative diagnosis of threatened abortion was made.

Past illnesses included rickets as an infant and measles as a child. Her menarche was at age 13; her periods lasted three to four days and occurred every 38 days. Dysmenorrhea, low back pain and abdominal cramps were present with almost every period, occasionally confining her to bed.

She was married in 1961 and in August of that year had an unusually heavy period and was thought to have had a miscarriage. She missed her December period and was hospitalized with pelvic inflammatory disease. At that time her pregnancy test was positive. From then until April 17, she had normal periods.

When first seen in June she had bright vaginal bleeding and generalized abdominal tenderness, but no rebound pain. Hemoglobin was 14.0 g. %, urinalysis normal, VDRL test negative, Aschheim-Zondek test positive. On June 14 she passed friable tissue, reported histologically as products of conception. Because of continued bleeding an examination under anesthesia was performed and a complete vaginal septum, two cervices and two uteri were found. The uteri were joined in the midline, and the right cervix appeared nulliparous. Both cervices were dilated and curettage was performed. The uterine cavities each measured two and a half inches in length. Curettings from the left cavity were reported as secretory endometrium heavily infiltrated with acute inflammatory cells; curettings from the right side were similar, and degenerated decidual tissue was recognized. The impression was that the patient had a uterus didelphys with a double vagina. She was referred to the Victoria General Hospital, Halifax, for further evaluation.

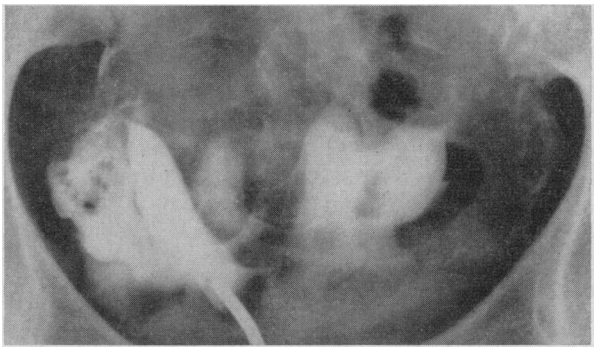

Fig. 1.—Hysterosalpingogram, right side.

At the Victoria General Hospital a hysterosalpingogram confirmed the presence of two uteri. The examinations showed the right vagina, cervix and uterine cavity (Fig. 1) to be smaller than the left (Fig. 2), which were considered normal in size. An intravenous pyelogram (IVP) did not show any anomaly of the urinary tract.

Consultation with members of the Department of Gynecology, Victoria General Hospital, led to the

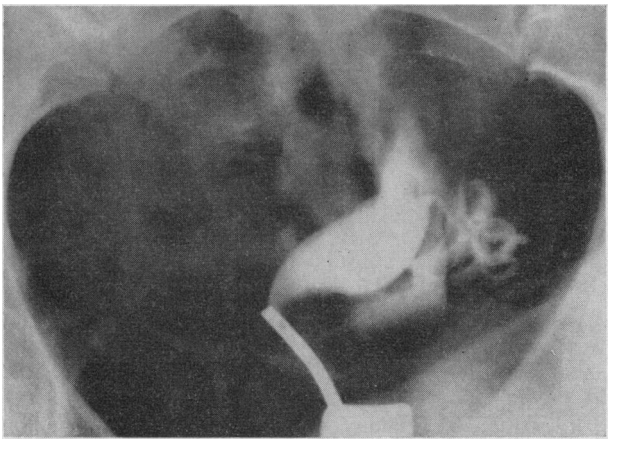

Fig. 2.—Hysterosalpingogram, left side.

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conclusion that reconstructive surgery should be postponed, because the patient was young and the history of one miscarriage was not necessarily an indication that she would be a habitual "aborter". The matter was discussed with the patient and her husband; it was suggested that since the left genital tract appeared to be larger and appeared more mature, the utilization of that side for coitus might result in a more successful pregnancy.

The patient was next seen on October 2, 1962, complaining of amenorrhea since August 20, "morning nausea", tension and nervousness. On October 30, she was thought to have an episode of viral influenza with nausea, abdominal cramps and leg pains. She also had postcoital dysuria. On November 26, she was hospitalized with lower abdominal pain, vaginal bleeding and tenderness over the uterus, which was estimated on abdominal examination to be of normal size. She was treated as a threatened abortion and her symptoms subsided in three days. Pelvic examination on December 5, 1962, revealed a pregnant left uterus. Further abdominal pains occurred during December and January. On January 3 quickening occurred. On seven occasions in February and March she reported dark red or brownish blood and some "tissue".

At 11 p.m. on May 1, 1963, labour began and she was admitted to hospital four hours later with uterine contractions occurring every 10 minutes. There had been no "show", but seeping of a little fluid was noted. The baby was felt to be in a left occipital anterior position, with a fetal heart rate of 140 per minute. At 10:00 a.m. on May 2 the left cervix was dilated 3 cm. with the presenting part 2½ cm. above the spines. The right uterus was in the pelvis, the cervix soft, undilated. Hard contractions were occurring every two minutes. This pattern continued throughout the day, and at midnight she was given sedation and intravenous fluids. At 8:00 a.m. on May 3 the cervix was three-quarters dilated but the right uterus was apparently preventing further progress through the pelvis. The membranes were ruptured but still no further progress was made. Laparotomy for cesarean section was started. At laparotomy the two uterine fundi were found to be totally separated and were joined at the cervical level only. Only one tube and one ovary were attached to each uterus. A female infant weighing 5 lbs. 3½ oz. was delivered by a low-segment transverse incision from the left uterus. The right uterus was four to five times normal size and could be pulled up, out of the pelvis, but when released would immediately drop into the pelvis again.

The postoperative course was uneventful except for tachycardia and slight fever on the fourth day. The protein-bound iodine and total serum iodine levels were normal. The baby exhibited no abnormalities or deformities. At time of discharge from the hospital she weighed 4 lbs. 15 oz.

When seen again on January 9, 1964, the patient gave November 26, 1963, as the date of her last normal menstrual period. She stated that she did have intercourse in the right vagina once, otherwise in the left vagina. Examination revealed the right fundus to be softer than the left and pulsating; the estimated date of confinement was October 2, 1964. A Papanicolaou smear was reported as Class II, some atypical cells having been noted. Her pregnancy was uneventful except for leg cramps which responded to calcium lactate. On September 21, 1964, she went into labour and after being in labour for eight hours she arrived at the hospital. She was having much discomfort but there was little progress of the baby, which was held up by the non-pregnant uterus lying in the pelvis (Fig. 3). The cervix was dilated to two fingers, the vertex presenting with the head low in the pelvis. A cesarean section was done through a well-thinned lower segment of the right uterus. She was delivered of a female infant weighing 4 lbs. 10½ oz. On September 25, 1964, it was necessary to do an exchange transfusion on the infant (because of Rh incompatibility), and a smaller transfusion was required on October 8. The patient was discharged on October 3, doing well. The baby was discharged on October 18, weighing 5 lbs. and doing well. Both children will be observed for evidence of abnormalities.

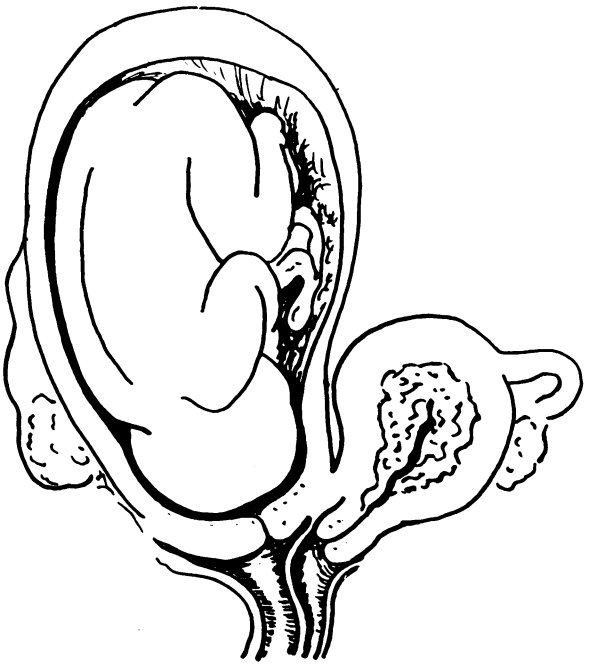

Fig. 3.—Diagram of uteri at term.

Contraception is another problem in this patient. She took contraceptive tablets from December 1964 to December 1965, at which time the tablets were discontinued. The patient developed a radial nerve paralysis, and although there was no evidence of association with the drug, it was considered wise to discontinue the medication. Subsequently she had a Gynekoil (small size) inserted into each uterine cavity. When the patient wants to become pregnant again, it will be of interest to see if pregnancy is possible in one uterus if just one Gynekoil is removed. When seen in July 1966, the patient was well.

DISCUSSION

In this patient the anomaly was that of double vagina and uterus. Normally the uterus and vagina develop embryologically by a fusion of the paired müllerian ducts, the fusion taking place from below upward. Anomalies result from an arrest of this process. In double vagina and uterus didelphys, as in this patient, practically no fusion takes place (Fig. 4). In 36 cases of uterine anomalies studied by Wilson and Harris[2] this situation was found in only four cases.

As might be anticipated, urinary tract anomalies are commonly associated with uterine abnormalities. Of 150 cases studied by Jones,[4] eight also had renal anomalies. Kamm and Beernik[3] reported a 5 to 10% incidence of associated anomalies of the urinary tract. Our patient had a normal IVP.

The abnormalities are usually not detected until pregnancy occurs. In Blair's[5] series in Glasgow the anomalies in 68% were unrecognized until the patient was actually in labour. They are usually associated with increased fetal loss. The patient often has a history of repeated abortions, and up to 38%[2] of first pregnancies in such cases end in abortions. There is a fetal loss of 25 to 45%.[3] In women with uterine anomalies prematurity is encountered two and one-half times as frequently as in women with normal uteri, and malpresentation occurs in 25%[3] to 56.8%.[5] Postpartum hemorrhage and retained placenta are encountered much more frequently than usual. However, this occurs more frequently with arcuate or bicornuate uteri than with uterus didelphys.

Before pregnancy, dysmenorrhea is a frequent problem. This patient had rather severe dysmenorrhea but did not seek medical attention. Her genital abnormalities were not discovered until she had become pregnant and had a miscarriage.

Twin pregnancy occurs more often in double uteri,[7] occurring about once in 12 pregnancies, which is approximately seven times the usual incidence. A peculiar feature, pointed out by Colaco,[8] of the radiograph of twin pregnancies in double uteri is that at no place do the fetuses overlap; it is as if they were "not on speaking terms".

As with most rare conditions in medicine, a high index of suspicion is needed to make the diagnosis early; and early diagnosis is necessary if the physician is to be prepared for the proper management of the problem. The occurrence of other abnormalities and of symptoms such as repeated abortions, prematurity and malpresentation should alert the physician to consider the possibility of uterine anomalies.

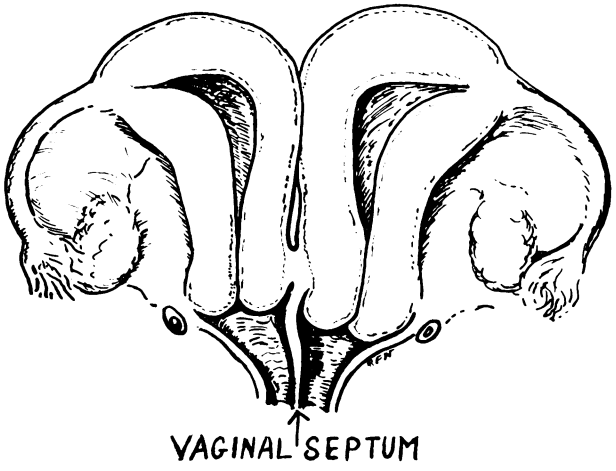

Fig. 4.—Diagram of patient's uterine and vaginal anomaly.

SUMMARY

A 21-year-old woman had a complete duplication of uterus, cervix and vagina, with the two uterine fundi completely separated. With the history of at least one abortion, complete investigation followed, and she was advised to become pregnant in the left uterus, by normal intercourse. This she did. She required extra care prenatally and frequent visits with supportive therapy. She had repeated episodes of vaginal bleeding, most likely from the non-pregnant uterus, and was treated by bed rest. However, she went to term and had a prolonged, difficult labour because the non-pregnant right uterus was impacted in the pelvis and caused obstruction. Because of this impaction she felt rectal pressure from the earliest stage of labour. She had a low-segment cesarean section on the left uterus. On the eleventh postoperative day both mother and child were discharged, doing well. No other anomalies were observed in either mother or child.

The patient had a subsequent pregnancy in the right uterus. This time the labour was obstructed by the non-pregnant left uterus, impacted in the pelvis. Again she had a cesarean section, this time of the right uterus. Both mother and child are doing well; the second child, however, required exchange transfusions.

In this particular case, it was possible for each fundus to produce a normal child in subsequent pregnancies.

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